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Case Report

Peripartum cardiomyopathy with biventricular thrombus which led to massive cerebral embolism

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ABSTRACT

A 37-year-old female who delivered her second child via a cesarean section 4 months previously presented to our hospital with gradual worsening of dyspnea on effort. Chest radiographic appearance showed cardiomegaly (cardiothoracic ratio 61%) and slight bilateral pulmonary congestion. Echocardiogram revealed diffuse hypokinesis of both left and right ventricles (left ventricular ejection fraction 29%) and large biventricular thrombus [left ventricular apex (28 mm × 21 mm, 22 mm × 14 mm) and right ventricular apex (16 mm × 11 mm)]. She was diagnosed as having peripartum cardiomyopathy (PPCM) and anticoagulation therapy was started. Surgical thrombectomy was not selected because of risk of complications. Massive cerebral infarction occurred 10 days after diagnosis. She was discharged with aphasia and right incomplete hemiplegia 65 days after admission.

Biventricular thrombus is a rare complication of PPCM. If high risk of massive embolism is considered, surgical thrombectomy may be warranted even in cases with low cardiac function.

<Learning objective: Biventricular thrombus is a rare complication of peripartum cardiomyopathy (PPCM). We report a case of biventricular thrombus secondary to PPCM. The decision to perform prophylactic surgical approach to ventricular thrombus is difficult in cases with low cardiac function.>

Introduction

Biventricular thrombus is a rare complication of peripartum cardiomyopathy (PPCM). The decision to perform a surgical approach to ventricular thrombus is difficult in cases with low cardiac function. We report a case of a Japanese female with biventricular thrombus secondary to PPCM.

Case report

A 37-year old Japanese female who delivered her second child via a cesarean section 4 months previously presented to our hospital with gradual worsening of dyspnea on effort. She had previous history of hypertension, type 2 diabetes mellitus, and obesity. During pregnancy, she had no dyspnea and the delivery was unremarkable. Her blood pressure was controlled around 130/80 mmHg without anti-hypertensive medication. Diabetes mellitus was controlled with insulin therapy only during pregnancy. Her peak body weight was 90 kg. She had no previous history of heart disease. Electrocardiogram (ECG) performed on second child delivery showed sinus tachycardia and no ST-T segment abnormality. Her second child was satisfactory. However, dyspnea on effort and chest discomfort occurred 2 months after delivery. On admission, her height was 165 cm and body weight was 94 kg (body mass index 33.3). She had blood pressure of 96/78 mmHg and heart rate of 100 bpm. Chest radiographic appearance revealed cardiomegaly (cardiothoracic ratio 61%) and slight bilateral pulmonary congestion. ECG showed sinus tachycardia with negative T wave in II, III, aVF, V6, and poor R progression in chest leads (Fig. 1). Blood test showed the level of B-type natriuretic peptide increased to 1015 pg/ml. Echocardiogram (UCG) revealed left ventricular (LV) dilatation and severe dysfunction [LV diastolic dimension (LVDd) 65 mm, ejection fraction (EF) 29%] (Fig. 2C). Not only LV but right ventricular (RV) wall motion also showed diffuse severe hypokinesis. There were two large thrombi in the LV apex (28 mm × 21 mm, 22 mm × 14 mm)

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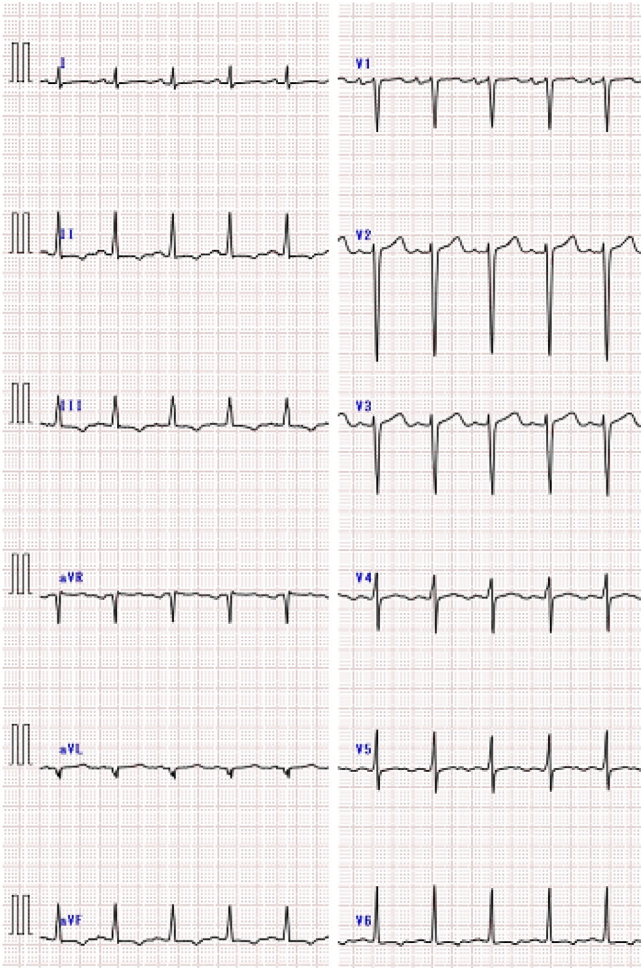

Fig. 1. Electrocardiogram on admission. Heart rate was 100 bpm. Negative T wave in II, III, aVF, V6 lead and poor R progression were seen.

and one in the RV apex (16 mm × 11 mm) (Fig. 2A and B). LV and RV thrombi were mobile. Cardiac computed tomography (CT) images showed no significant coronary artery stenosis. She was diagnosed as having congestive heart failure caused by PPCM. Angiotensin-converting enzyme inhibitors (ACEI) and diuretics were started. Surgical thrombectomy was considered. However, left ventriculotomy was considered to have a high risk in this patient, because this would lead to further LV dysfunction. Thrombus resection via aortic valve retrogradely or via mitral valve with left atriotomy was an alternative way, but complete thrombus resection may not be attained, and this procedure may be associated with a risk of embolization. After discussion with cardiac surgeons, conventional anticoagulation therapy was selected. We started continuous intravenous injection of heparin with oral administration of warfarin. Activated partial thromboplastin time (APTT) was controlled at around 45–60 s. Thrombolytic therapy was not selected because it may have increased the risk of embolization.

Heart failure gradually improved. In UCG at day 8, ventricular thrombus became smaller (LV 20 mm × 17 mm, 19 mm × 13 mm, RV 11 mm × 8 mm), however the mobility of thrombus was enhanced. Bilateral ventricular wall motion was not improved at all. On day 10, her consciousness level suddenly deteriorated to Glasgow Coma Scale 8 points (E1V2M5). Right complete hemiplegia occurred. Emergent contrast enhanced CT image revealed total occlusion of left common carotid artery with thrombus (Fig. 3A). At that time, APTT was 54.9 s and prothrombin time and international normalized ratio was 1.54. Catheter thrombectomy was performed. UCG after the procedure revealed disappearance of biventricular thrombus. Although RV thrombus disappeared, apparent acute pulmonary thromboembolism was not seen during the clinical course.

Because midline shift in brain CT image was seen, decompressive craniectomy was performed to prevent brain herniation at day 12 (Fig. 3B). We performed right heart catheterization in order to monitor perioperative hemodynamic status. Pulmonary artery pressure was 49/33 mmHg, mean pulmonary capillary wedge pressure was 33 mmHg, RV pressure was 46/11 mmHg, mean right atrial pressure was 13 mmHg, and cardiac output and cardiac index were 5.71 and 2.77, respectively. We increased the amount of diuretics to prevent the worsening of heart failure. Her consciousness level gradually improved after the operation. We continued ACEI, diuretics, and anticoagulation therapy. Moreover, we added beta-blocker after heart failure was improved. She was discharged with aphasia and right incomplete hemiplegia at day 65 after admission.

After 12 months, UCG showed LVDd 48 mm, and EF 62%. ECG was almost normal. Anticoagulation therapy was stopped, and no ventricular thrombus was seen in UCG at 13 months.

Discussion

PPCM is a form of heart failure that develops in the last month of pregnancy or within 5 months of delivery in patients without preexisting heart failure [1]. The incidence of PPCM varies between race and region, reported to be 1 per 3000–4000 child births in the USA, 1 per 1000 child births in South Africa, and 1 per 300 child births in Haiti [2–5]. In Japan, the incidence was reported as 1 per 20,000 child births in 2009 [6]. However, the incidence may be underestimated because the diagnosis of this rare disease is difficult.

The etiology of PPCM is still unknown. Some studies suggested that myocarditis from viral infection maybe the cause of PPCM [7,8]. Another possible mechanism is an autoimmune response against maternal myocardium provoked by release of fetal antigen into maternal blood [9]. On the other hand, the relationship between hemodynamic volume overload during pregnancy and occurrence of PPCM was previous reported [10]. The role of a 16 kDa prolactin derivative produced by proteolytic cleavage of prolactin secondary to unbalanced oxidative stress, which presents during late pregnancy and early postpartum period has been noted [11]. Medicines causing reduced secretion of prolactin from posterior pituitary gland or working as a D2 receptor antagonist such as bromocriptine have been reported to be effective in controlling PPCM patients [12].

Mortality of PPCM was reported to be significant lower than that in idiopathic cardiomyopathy [13]. LVDd > 56 mm at the time of diagnosis, EF < 45% at two months after the disease occurrence, existence of LV thrombus at the time of diagnosis, and being African-American have been reported as factors of poor prognosis [14].

Because of high procoagulant activity in the postpartum period due to elevation of factors VII, VIII, X, fibrinogen, and von Willebrand factor, PPCM patients who have low cardiac function have high risk of ventricular thrombus, which is estimated to be 10–30% [14–16]. In PPCM patients, especially with low cardiac EF, anticoagulation therapy is strongly recommended.

Biventricular thrombi associated with PPCM are quite rare. To date, only 6 cases have been reported. In these reports, only anticoagulation therapy was prescribed and prophylactic surgical thrombectomy was not performed. Thromboembolism occurred in 2 cases. One case suffered cerebral infarction and was treated with recombinant tissue plasminogen activator. Another case suffered acute saddle embolism and was treated with surgical resection [17–22]. Although acute pulmonary artery thromboembolism was not reported in these 6 cases, it may develop in patients with PPCM

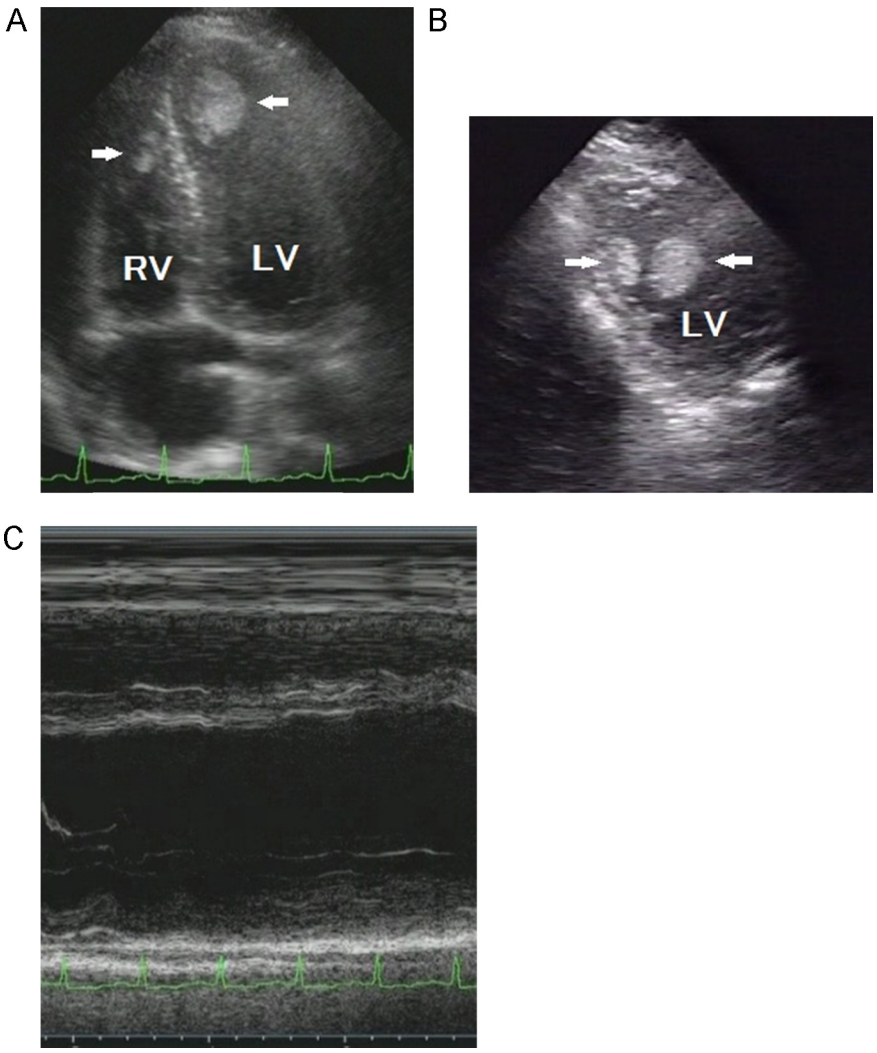

Fig. 2. Transthoracic echocardiogram on admission. (A) An apical 4-chamber view of transthoracic echocardiogram on admission. (B) A short-axis view of transthoracic echocardiogram on admission. These images revealed massive thrombus in left ventricular apex (28 mm × 21 mm, 22 mm × 14 mm) and right ventricular apex (16 mm × 11 mm). (C) M-mode echocardiogram on admission. Left ventricular diastolic dimension was 65 mm, and ejection fraction was 29%. LV, left ventricle; RV, right ventricle; Arrow, thrombus.

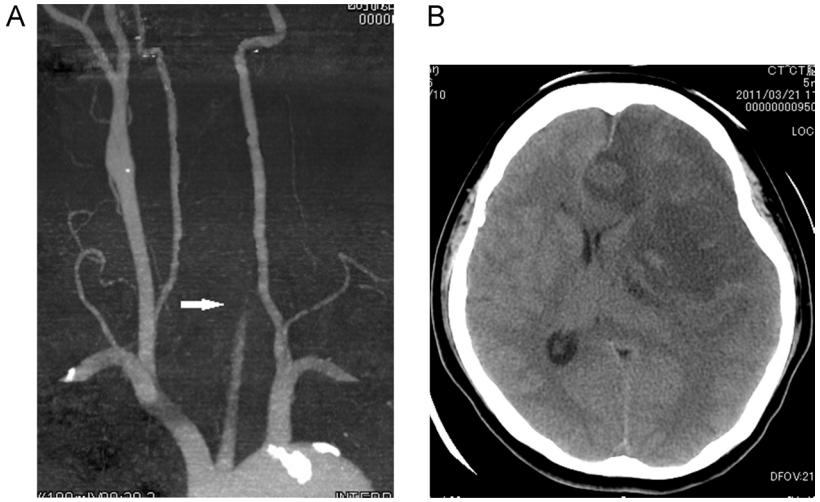

Fig. 3. Computed tomography (CT) images when massive cerebral infarction occurred. (A) A reconstructed contrast enhanced CT image of cervical area just after thrombus embolization of left carotid artery. Left common carotid artery was completely occluded (arrow). (B) A brain CT image two days after cerebral infarction. Midline shift was revealed and decompressive craniectomy was performed.

and RV thrombus. In PPCM, embolism to cerebral artery, coronary artery, splenic artery, and artery of lower extremity has been reported [21–24].

Amos et al. reported that in 55 patients, the incidence of LV thrombus complicating PPCM was 17%, and only one patient suffered thromboembolism (coronary artery thromboembolism) [14]. Napporn et al. reported the incidence of LV thrombus complicating PPCM was 10% in 58 patients, and there were no patients who suffered thromboembolism [16]. There is no consensus or clinical guidelines to outline the best management strategy for ventricular thrombus due to PPCM. Anticoagulation therapy was reported to be effective in most of the cases. Deterioration of ventricular thrombus in the early phase of the disease has possibility of embolization with the improvement of ventricular wall motion. However, in this case, when cerebral embolization occurred, biventricular wall motion was not improved at all.

The strategy of surgical resection may be justified only in cases which large and mobile thrombus. However, cardiac ventriculotomy will make further decrease in systolic function, especially if the LV function is already poor. In addition, ventriculotomy may potentially induce ventricular arrhythmia. Also, the risk of hemorrhagic complication is high under anticoagulation therapy. An alternative way to remove ventricular thrombus is resection via aortic valve retrogradely or via mitral valve with right-sided left atriotomy. However, these procedures cannot guarantee total thrombus resection and there exists considerable risk of thromboembolism during the procedure. All surgical procedures to remove thrombus need cardiac arrest with cardiopulmonary bypass support. Low cardiac output syndrome after operation will inevitably occur in patients with LVEF around 20% and uncontrolled heart failure. In a previous study, EF < 30%, uncontrolled congestive heart failure, and urgent operation were determinants of operative mortality for adult patients undergoing open heart surgery [25].

A case of surgical ventricular thrombectomy in PPCM was reported in 2008 from a Japanese institute [26]. A 32-year-old PPCM patient who had large and mobile LV thrombus (35 mm × 20 mm) with low EF (20%) underwent thrombus resection through a right-sided left atriotomy, with cardiopulmonary bypass. Although the thrombus was successfully removed, UCG revealed new left apical ventricular thrombus at postoperative day 3. Because the thrombus was immobile, anticoagulation therapy successfully eliminated the new thrombus without any clinical embolic event. We could not find other reports in which prophylactic surgical resection of ventricular thrombus due to PPCM was performed. In our case, although the patient had high risk of thromboembolism, we selected conventional medication strategy, considering severe complications from surgical operation. This resulted in massive cerebral embolism.

In conclusion, we experienced a rare case of biventricular thrombus associated with PPCM. Aggressive surgical thrombectomy may be warranted in PPCM cases with large mobile left ventricular thrombus, even in patients with poor cardiac function.

Conflict of interest

Authors declare no conflict of interest.

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